

D. KENWIN HARRIS AND W. G. F. ADAMS: ACRO-OSTEOLYSIS FROM POLYMERIZATION OF VINYL CHLORIDE



FIG. 1.—Case 1. Erosions of the terminal phalanges of index and middle fingers, both hands.

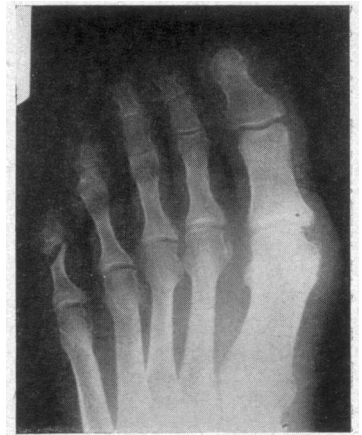


FIG. 2.—Case 1. Disorganization of the fifth proximal interphalangeal joint and erosions of the distal ends of the fifth proximal phalanges of both feet.

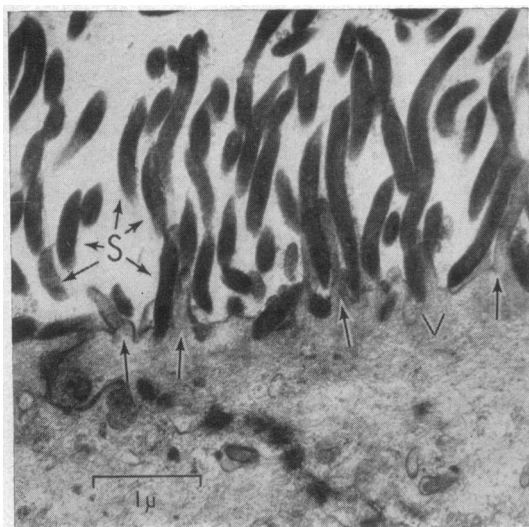


FIG. 3.—Case 2. Osteolysis is present in all the terminal phalanges except the left ring finger.



FIG. 4.—Case 2. Cortical erosion of posterior surface of the patella.

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INTESTINAL SPIROCHAETOSIS



Surface of two epithelial cells with numerous spirochaetes (S) orientated in the long axis of the cell. Arrows indicate microvilli. V is a reactive vacuole.

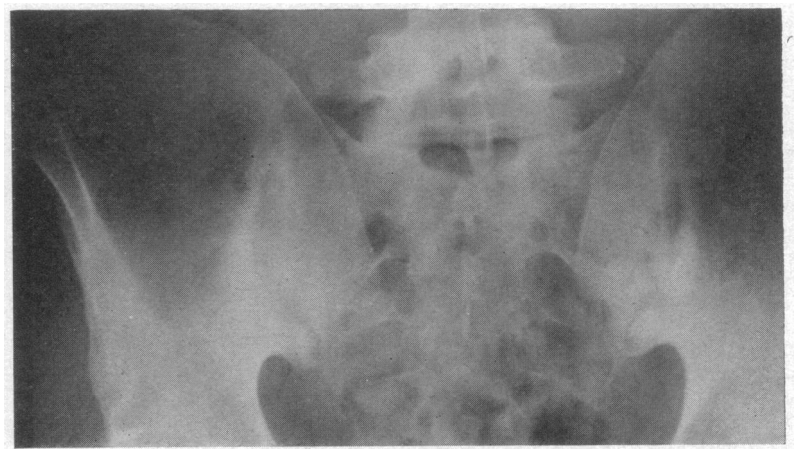


FIG. 5.—Case 2. Widening of the sacroiliac joint space, with cystic marginal sclerosis.

Preliminary Communications

Intestinal Spirochaetosis

[WITH SPECIAL PLATE BETWEEN PAGES 708 AND 709]

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The purpose of this paper is to draw attention to a form of intestinal spirochaetosis that does not appear to have been previously described and yet would seem to be not infrequent. The condition was first noticed while the rectal mucus membrane was being viewed through the electron microscope.

INITIAL OBSERVATIONS

The patient, a man aged 64, had complained of persistent diarrhoea for three years. A rectal biopsy was taken as part of the clinical investigation, and on light microscopy nothing of note was seen. Small fragments of this biopsy specimen were fixed in 1% osmium tetroxide in phosphate buffer at pH 7.4 and embedded in Araldite, and thin sections were viewed in a Siemens Elmiskop 1A electron microscope.

Clinical Features of 10 Cases of Intestinal Spirochaetosis

Case No.	Sex	Age	Complaint	Duration of Symptoms	Clinical Diagnosis
1	M	64	Diarrhoea	3 years	?
2	M	52	Mucoid diarrhoea	2-3 days	Carcinoma of colon
3	M	49	Diarrhoea	4 weeks	?
4	F	19	Abdominal pain	1½ years	Appendicitis.
5	F	64	Constipation	5 "	Haemorrhoids
6	M	56	Rectal bleeding	6 months	Carcinoma of colon
7	M	56	"	8 "	Haemorrhoids
8	M	60	Intermittent rectal bleeding	1 year	Carcinoma of colon
9	M	61	Rectal bleeding	2 years	Multiple polyps
10	M	63	Diarrhoea	2 weeks	Carcinoma of colon

The electron microscopical findings are illustrated in the Fig. (Special Plate). Adherent to the surface of the epithelium was a forest of organisms, orientated in the long axis of the cells. The organisms were short and thin and had two or three short curves. They measured up to 3 μ in length and only 0.15 μ in average diameter. They were too short and thin for bacilli. Under high magnification occasional organisms, cut tangentially, showed bunches of spiral filaments. This finding, together with the shape and size of the organisms, indicated that they were spirochaetes.

The spirochaetes sometimes penetrated short distances into the epithelial cells, pushing the plasma membrane before them. Occasionally a vacuole could be seen forming in the cytoplasm of the cell at the tip of the organism. Some of the epithelial cells appeared to have an increased number of lysosomes, but no other reaction was noticed. There was no significant inflammation. Where the surface of the cells had been cut transversely each organism could be seen to be surrounded by a cluster of microvilli. This probably accounts for the consistent longitudinal orientation of the organisms.

Reappraisal of the routine histological sections in the light of the electron microscopical findings revealed a very faint blue haze on the surface of the mucosal cells.

SUBSEQUENT INVESTIGATIONS

It was soon realized that this basophil reaction had been seen histologically in other rectal biopsies but that it had been inter-

preted as an unusual staining of the brush border of no significance. A review of 100 consecutive rectal biopsies from the files of this department was then undertaken. In nine cases a faint blue haze, exactly as seen in the first biopsy, was present. It was clearly necessary to find out if all of these cases represented a single entity. Small blocks of tissue were excised from the paraffin wax in which they had been stored, the wax was removed in chloroform, and the tissue was post-fixed in osmium tetroxide, after rehydration through graded alcohols. Thin sections were then examined in the electron microscope.

Preservation of the tissue was naturally poor, but in all nine cases spiral organisms identical in size and shape to those seen in the original case were readily identified. As before, they were closely applied to the surface epithelium and in some places just penetrated the cell surface.

It was concluded that all these cases represent a single pathological entity, which we have called intestinal spirochaetosis. This condition can be readily diagnosed in rectal biopsies. It is recognized histologically, even with the low-power objective, as a distinct haematoxyphilic zone occupying the site of the normal brush border. This zone is about 3 μ in width. It is not possible to resolve individual organisms in routine paraffin sections because of the thickness of the sections, but individual organisms can be resolved by the light microscope in very thin (1 μ) sections prepared from Araldite-embedded material.

CLINICAL OBSERVATIONS

The main clinical features of the 10 cases are shown in the Table. Both sexes are involved and there is a wide age distribution. All patients had symptoms referable to the bowel. In five cases carcinoma of the colon was definitely diagnosed either by the first biopsy or at a later date. It is of interest that in these cases only the normal mucosa, adjacent to the cancer, was infested by spirochaetes. In three cases haemorrhoids were thought to be the cause of symptoms.

There is evidence that intestinal spirochaetosis can persist for considerable periods. In Case 2, for example, the condition was diagnosed in two biopsies taken 17 days apart, but after preoperative antibiotic treatment spirochaetes were not identified in the resection specimen. In Case 1 two biopsies were both positive; they were separated by a period of five months, during which the patient's symptoms persisted. Case 8 had five biopsies in six years and all showed clear evidence of spirochaetosis.

Case 4 is of special interest. This patient had had attacks of abdominal pain for one and a half years which led to appendectomy. She still had symptoms, and haemorrhoids were injected. Spirochaetosis was present not only in the rectal biopsy specimen but also in the appendix removed two months earlier.

DISCUSSION

Since the last century spirochaetes have been known to be present in the intestinal tract, but have usually been regarded as harmless commensals. However, there are scattered reports in the literature that large numbers of spirochaetes can be found in the stool of patients with various forms of dysentery and cholera (Parr, 1923). In addition there are dysentery-like diseases, associated with spirochaetes, which have responded only to treatment with arsenicals (Sáenz, 1925). It is also claimed that spirochaetes are responsible for guinea-pig diarrhoea (Sanarelli, 1927).

Presumably because they have been considered to be non-pathogenic, there has been little attempt to culture and further classify these organisms. However, the morphological descriptions of the commonest organisms, the so-called *Spirochaeta eurygyrata* (Hogue, 1922), closely resemble the spirochaete found in this group of patients. Because these organisms stain with haematoxylin, we think that they would be best classified as *Borrelia*. In future publications further details will be presented.

It is not possible at the present time to decide the significance of our findings. All the patients had rectal biopsies performed because of symptoms of one sort or another referable to the bowel. Some were found to be suffering from carcinoma of the colon. On the other hand, others had dysenteric symptoms of unknown cause, and these might possibly be cases of "spirochaetal dysentery." Antibiotic treatment of some of these cases may provide an answer to this problem, since we have reason to believe that the spirochaetes disappeared in one case after preoperative antibiotic therapy. There is also the possibility that this form of spirochaetosis gives rise to vague abdominal symptoms simulating appendicitis. It remains quite possible, however, that this attachment of spirochaetes to the surface of the colonic mucous membrane has no clinical significance.

SUMMARY

Ten cases of a new and apparently common form of intestinal spirochaetosis are described. The condition was first recognized by electron microscopy, but diagnosis can be made on routine histological preparations. Short spirochaetes, tentatively classified as *Borrelia eurygyrata*, are attached to and penetrate short distances into the surface epithelial cells. They are characteristically orientated in the long axis of the cells. At present it is not possible to decide the significance of our findings.

We acknowledge the invaluable technical assistance of Mr. D. McSeveney, whose skill made this study possible; we thank Mr. D. H. Clark, who referred the first patient to us; and we acknowledge the financial assistance from the M.R.C., who provided the electron microscope.

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Medical Memoranda

Volvulus of the Small Intestine after
Herniorrhaphy

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Primary volvulus of the small intestine is rare in Great Britain and its cause is usually obscure. Renton (1965) found five reported examples and added one of his own. Secondary volvulus is relatively common and its cause is better understood. The present case is reported in the hope that it may contribute to further understanding of the factors precipitating intestinal volvulus.

CASE REPORT

At 2 p.m. on 15 March 1966 a 68-year-old farmer developed lower abdominal pain after lifting a heavy weight while working in his fields. At the same time he noticed a tender swelling in his right groin. He was admitted to hospital at 6 p.m. For two years he had been aware of a small hernia in the right groin which previously had always been easily reduced. During the last seven years he had experienced attacks of abdominal distension, associated with flatulent discomfort, which lasted up to 10 days and resolved with the passage of watery diarrhoea. These attacks occurred two or three times a year and appeared to be unconnected with any disturbance in his hernia. He was not a vegetarian and ate a regular balanced diet.

On admission his general condition appeared satisfactory. His pulse was 72. The blood pressure was 160/80 mm. Hg. His chest showed signs of chronic bronchitis. A tender irreducible right inguinal hernia was present. The abdomen was not distended but bowel sounds were increased.

That evening the inguinal hernia was explored (N.H.V.) under general anaesthesia. It was found to be direct in type and to contain 6 in. (15 cm.) of gangrenous ileum, which was resected. End-to-end anastomosis was carried out and the hernia was repaired. Over the next few days his abdomen became painlessly distended and he was thought to have developed a paralytic ileus. On 19

March his bowels moved loosely and thereafter he had two or three loose motions daily, but the abdominal distension remained. He vomited 200 ml. of stomach contents on 22 March. When this was repeated on the 23rd an intravenous infusion of dextrose saline was started. On the 24th his abdomen suddenly became further distended, having increased 2 in. (5 cm.) in girth overnight. There was generalized abdominal tenderness and guarding. His pulse was 100. The blood pressure was 135/80. X-ray examination of the abdomen showed small-bowel distension, with fluid levels suggestive of obstruction in the distal ileum.

After the passage of an oesophageal tube and aspiration of the stomach, laparotomy was performed (C.J.C.R.). Blood-stained fluid was present in the peritoneal cavity. The small intestine was very distended, was dark purple in colour, and its viability appeared doubtful. The entire small intestine, except for the proximal 4 ft. (1.2 m.) of jejunum, had undergone a clockwise rotation of 720 degrees. There were no adhesions, and the previous anastomosis was patent and lying free in the right iliac fossa. No congenital or acquired abnormality was found to account for the volvulus. On reducing the volvulus the circulation to the intestine recovered. Because of the gross distension the bowel was deflated by a suction tube introduced through an enterotomy. The bowel was closed and the original anastomosis, which had leaked after handling, was oversewn. Later the same evening sputum retention and bronchospasm produced increasing dyspnoea, cyanosis, and carbon dioxide retention, leading to respiratory failure. Tracheostomy was performed (N.H.V.), allowing regular bronchial toilet. With physiotherapy, antispasmodics, and antibiotics respiratory function was improved. Gastric suction and intravenous infusions were continued until 28 March, when the abdominal distension started to resolve. His bowels thereafter moved once a day. Two episodes of vomiting on 2 and 6 April necessitated further gastric suction. By 8 April the vomiting and the abdominal distension had completely settled. The tracheostomy tube was removed and convalescence continued uneventfully.

COMMENT

In primary volvulus of the small intestine there is no obvious predisposing cause. It is commoner in Africa and India, where it is associated with a vegetarian diet of bulky indigestible